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Case Report

Intramural florid cystic endosalpingiosis of the uterus: A case report and review of the literature

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ABSTRACT

Objective: We report a case of intramural florid cystic endosalpingiosis in the lower uterine segment of the uterus.
Case report: A 43-year-old female presented with vaginal bleeding. Abdominal computed tomography suggested a leiomyoma with cystic degeneration. A total hysterectomy revealed a 4.0 cm × 3.8 cm cystic mass in the lower uterine segment. The cystic space microscopically was lined with a single layer or stratified layer of ciliated columnar cells that resembled tubal epithelium without cytologic atypia. The glandular spaces were surrounded by normal myometrium with no evidence of periglandular endometrial stroma, which was consistent with the diagnosis of florid cystic endosalpingiosis.
Conclusion: Florid cystic endosalpingiosis involving the uterus is a rare and clinically unexpected finding; however, it should be considered in the differential diagnosis of a uterine mass.

Introduction

Endosalpingiosis in women is characterized by benign glands lined by tubal type epithelium, and involves the peritoneum, subperitoneal tissues, and retroperitoneal lymph nodes [1,2]. It is nearly always asymptomatic and is primarily detected incidentally during microscopic examination. Florid cystic endosalpingiosis (FCE) is a rare form of endosalpingiosis and presents as a tumor-like lesion; its uterine involvement is even rarer, with fewer than 17 cases published to date [1–14]. We report a case of intramural FCE in the lower uterine segment of the uterus.

Case report

A 43-year-old woman, parity 1, 0, 0, 1, presented with vaginal bleeding. Abdominal computed tomography revealed a 4.0 cm × 3.8 cm ill-defined heterogeneously dense lesion on the left side of the lower uterine segment (Fig. 1). The lesion displayed internal cystic change, which suggested a leiomyoma with cystic degeneration. The patient underwent a total hysterectomy. A 4.0 cm × 3.8 cm mass was in the lower uterine segment. The mass was relatively well circumscribed and white. A cut section revealed variously sized cystic formations without any communication with the endometrial cavity (Fig. 2). The mass microscopically was extensively involved by numerous glands that were lined with a single or stratified layer of ciliated columnar cells resembling tubal epithelium. There was no cytologic atypia (Fig. 3). The glandular spaces varied widely from small round glands to markedly distended large spaces with folding. These glands were surrounded by normal myometrium with no evidence of periglandular endometrial stroma.

Discussion

Endosalpingiosis is one aspect of the triad of non-neoplastic secondary Müllerian lesions with a pure or dominant glandular component; the other aspects of the triad are endocervicosis and endometriosis [1,6]. Endosalpingiosis is mostly an asymptomatic lesion with no serious prognostic association [15]. However, it may rarely present as a tumor-like lesion. Florid cystic endosalpingiosis has been reported in various locations such as the uterine cervix and corpus, ovaries, fallopian tubes, paraovarian area, urinary bladder, ureter, spleen, colon, and appendix [1,16–19]. Florid cystic endosalpingiosis involving the uterus is rare— only 16 cases have been reported in the literature (Table 1) [1–14]. The median age of

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Fig. 1. The abdominal computed tomography image reveals a 4.0 cm × 3.8 cm ill-defined heterogeneously dense lesion on the left side of the lower uterine segment. An internal cystic change (arrows) suggests a leiomyoma with cystic degeneration.

Fig. 2. The gross findings. A relatively well circumscribed multicystic mass (★) is in the lower uterine segment. There is no communication between the cystic spaces and the endometrial cavity (arrowhead).

Fig. 3. The microscopic findings. Numerous cystic spaces (C) are within the myometrium (M) [hematoxylin and eosin (H & E); magnification, ×1.25]. The peri-glandular endometrial stroma is absent. The cystic spaces are lined with a single layer or stratified layer of ciliated columnar cells resembling tubal epithelium (arrow) (inlet). (H & E; magnification, ×400).

Table 1
Previously reported cases of uterine florid cystic endosalpingiosis.

Case	References	Age	Clinical presentation	Location	Gross findings	Size	Clinical impression
1	Bryce et al, 1982 [3]	25	Incidental finding on cesarean section	S	Multiple cysts	Up to 1.0 cm	NR
2	Clement and Young, 1999 [1]	20	Incidental finding on cesarean section	S	Multiple "cystic blebs"	NR	NR
3	Clement and Young, 1999 [1]	41	Menometrorrhagia and dyspareunia	I	Numerous cysts with a spongy appearance	NR	Leiomyoma
4	Clement and Young, 1999 [1]	43	Menometrorrhagia and pelvic pain	I	Multiple cysts	Up to 3.2 cm	Cervical cancer was suspected because of the enlarged cervix
5	Heatley and Russell, 2001 [4]	73	Lower abdominal swelling	I	Multicystic nodules	Up to 1.6 cm	NR
6	Chang et al, 2003 [5]	45	Abdominal dull pain and irregular menstruation	S	Multilocular cyst	8.2 cm × 7.5 cm × 6.0 cm	Adnexal tumor (radiologically) Degenerative cyst of a myoma (during surgery)
7	Fukunaga, 2004 [2]	51	Lower abdominal pain	S	Mass with cysts	Up to 4.0 cm	Cystic ovarian tumor
8	Kajo et al, 2005 [6]	50	Palpable mass	I	Tumor-shaped configuration	4.5 cm	Adnexal tumor
9	Lee et al, 2005 [7]	52	Pelvic mass incidentally detected on check up	S	An oval cystic mass and a solid mass	13 cm and 8.5 cm in diameter, respectively	Cystic ovarian tumor (radiologically), leiomyoma with cystic degeneration (during surgery)
10	Youssef et al, 2006 [8]	49	Menorrhagia	S	Multiple cysts	Up to 2.0 cm	NR
11	Cil et al, 2008 [9]	45	Metrorrhagia, pelvic pain, continuous vaginal bleeding	I	Unilocular cyst	3.5 cm	Cystic adenomyosis
12	Shim et al, 2008 [10]	54	Vaginal bleeding	S	Multicystic mass	Up to 6.0 cm × 4.5 cm × 4.5 cm	Bilateral benign cystic ovarian tumors
13	Suarez-Vilela et al, '2009 [11]	51	Menorrhagia	S	Several cysts	5.0 × 3.0 cm	Leiomyoma with cystic degeneration
14	Taneja et al, 2010 [12]	40	Pelvic pain, dysfunctional uterine bleeding	S	Irregular gray-white tissue	NR	Endometriosis (radiologically)
15	Patonay et al, 2011 [13]	44	Abdominal pain	S	Multiple vesicles	Up to 0.8 cm	NR
16	Resenberg et al, 2011 [14]	50	Recurrent pelvic illness and pain	S	Solid mass with a cystic center	9.5 cm × 7.0 cm × 6.0 cm	NR
17	Present case	43	Vaginal bleeding	I	Multilobular cyst	4.0 cm × 3.8 cm	Leiomyoma with cystic degeneration of

I = intramural; NR = not recorded; S = subserosal.

these 17 cases, which includes the present case, is 45 years (range, 20–73 years) and the median size in 14 recorded cases is 4.0 cm (range, 0.8–13.0 cm). The most common presenting symptoms are menometrorrhagia ($n = 5$) and abdominal pain ($n = 5$), followed by a palpable mass ($n = 2$), vaginal bleeding ($n = 2$), incidental finding on cesarean section ($n = 2$), and lower abdominal swelling ($n = 1$).

Florid cystic endosalpingiosis of the uterus can be categorized into two types, based on their location: intramural or subserosal. Five reported cases [1,4,6,9] and the present case are classified as the intramural type of FCE. Because of the intramural location, most of the mass is considered a leiomyoma with cystic degeneration or cystic adenomyosis. However, cervical cancer or cystic ovarian tumor has also been suggested. Enlargement of the cervix raised concerns for cervical cancer in one patient, and the patient underwent total hysterectomy with bilateral salpingo-oophorectomy and pelvic lymph node dissection. Frozen biopsy was performed with a diagnosis of "suspicious for minimal deviation adenocarcinoma" [1].

Eleven previously reported cases were classified as the subserosal type [1–3,5,7,8,10–14]. The masses were located on the anterior and posterior surfaces of the uterine fundus. Involvement of the ovarian surface [1,8] and extensive involvement of peritoneum [13] have also been reported. It is unusual for a FCE to occur inside a uterine subserosal leiomyoma [11]. The multicystic appearance of the FCE may simulate a cystic ovarian tumor, and the subserosal location of the mass can complicate distinguishing it as a true uterine lesion.

Microscopic differential diagnosis includes adenomyosis, adenomyoma, adenofibroma, atypical polypoid adenomyoma, florid mesonephric hyperplasia, and tubal metaplasia of the endocervix [1,2,6]. The lack of endometrial stroma in the present patient easily ruled out adenomyosis and adenomyoma. The multicystic mass-like appearance suggested an adenofibroma. The epithelial lining of an adenofibroma may be endometrioid, ciliated of the tubal type, or columnar mucinous of the endocervical type. However, the prominent tubal epithelium in the present case excluded the possibility of an adenofibroma and an atypical polypoid adenomyoma [2,6,20]. Tubal metaplasia of the endocervix was also excluded because the mass lesion had no communication with the endometrial cavity.

The pathogenesis of FCE is largely unknown; however, müllerianosis has been described wherein the coelomic epithelium lining of the peritoneal cavity may undergo changes towards primary Müllerian epithelium (including the tubal, endometrial, and endocervical epithelium) [10].

We report a rare case of FCE involving the uterus of a 43-year-old woman. This was a clinically unexpected finding. Being aware of the fact that endosalpingiosis can rarely form cystic mass-like lesions is important for clinicians and pathologists to make a correct diagnosis and prevent unnecessary surgical procedures. A full understanding of the macroscopic and microscopic features of this entity will facilitate making a correct diagnosis.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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